

# Pediatric plastic surgery research needs in global health: A scoping review

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## Abstract

**Introduction:** Plastic surgery in developing countries have focused on children for decades, however there is currently no formal literature review. This scoping review aimed to identify key themes and research gaps in global pediatric reconstructive surgical research to effectively guide future initiatives.

**Methods:** This was an IRB-approved scoping review of publications from EMBASE and MEDLINE between 2000 to 2013 relevant to pediatric plastic surgery in low-resource communities. Two independent investigators completed the primary review and three independent investigators completed the secondary review. Descriptive demographic data was collected.

**Results:** 106 studies were included. The majority came from Asia (57%), followed by Africa (40%) and Central/South America (11%). The clinical domains were: burns (n=50), cleft lip/palate (CLP, n=32), general plastic surgery (n=10), craniofacial (n=6), flaps and grafts (n=3), and hand surgery (n=1). Burn publications stressed burn prevention (64%) and public education (54%) as needs. CLP research identified a need for monitoring/evaluating current programs (19%) and increasing public awareness (19%). General plastic surgery research emphasized a need for environmental hazard prevention (60%). International agencies (n=25) invited further research in developing local healthcare infrastructure, and monitoring/evaluating current projects.

**Conclusions:** A systematic review is welcome in pediatric burns and CLP. Hand surgeries, flaps/grafts, and other craniofacial anomalies are significantly underreported. A paucity of clinical research, and research from Central/South America remains. Emphasis on developing local infrastructure and evaluating current international volunteer missions is welcome. The significant gaps remaining in global pediatric plastic/reconstructive surgical research constitute opportunities for future international projects.

## Introduction

Internationally, plastic surgery is making its mark as a necessary surgical specialty in Low and Middle Income Countries (LMICs). Plastic surgeons are uniquely qualified to address the burden of surgical disease in the developing world, most notably through management of burn injuries, facial congenital anomalies and traumatic injuries [1].

Children have been the focus for plastic surgeons who work in LMICs for decades. Early childhood age is associated with a disproportionately high incidence of burns, accidents and poorer outcomes [2-10]. The focus on children substantially improves outcomes for burns, traumatic injuries, and seriously debilitating congenital anomalies, notably cleft lip/palate (CLP) [11-15].

Despite the fact that resources were allocated for management of burns and congenital anomalies, the quality of care from foreign agencies is now a priority. Concerns with inappropriate follow-up, unsustainable funding, meeting the 'body count', i.e., operating on medically unfit children to meet a quota, significantly contributed to the movement towards surgical harm reduction. Since 2008, Operation Smile, Smile Train, and Resurge International were among the invested international non-governmental agencies that began to evaluate the quality of care provided [16-21].

While these examples are a clear indication of growing interest and research in this field, no formal review has been conducted. The objective of this scoping review is to identify key themes and research gaps in global pediatric reconstructive surgical research to effectively guide future initiatives.

## Methods

This study was reviewed and approved by the institutional review board of Queen's University in Ontario, Canada. A scoping review was selected as the best research methodology that would meet the study's broad objective of identifying dominant themes and research needs. Since pediatric plastic surgery research in LMICs is a heterogeneous field where no formal review had previously been conducted,

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this method was also applied because it provided the flexibility of modifying inclusion/exclusion criteria *post hoc*. A systematic review and meta-analysis was deemed to not be an appropriate fit for a broad, heterogeneous field where a formal assessment of study quality would not be pertinent to this study's objectives. To guide the scoping review, the framework designed by Arksey and O'Malley [19] and modified by Levac, et al. [20]. There were four stages to this review process: (1) Establishing the question (2) Identifying and selecting relevant literature (3) Charting the data, and (4) Collating, summarizing and reporting the results.

**Establishing the research question:** A team-based approach was used to determine a broad research question with a clearly articulated scope of pediatric plastic surgery in LMICs. From this, a search strategy was devised using the following key terms: Burns, Cleft lip and/or Cleft Palate (CLP), Craniofacial Anomalies, Flaps and Grafts, Hand Trauma, General plastic surgery, and International Agencies. General plastic surgery referred to articles that covered more than one clinical domain of plastic surgery.

**Identifying and selecting relevant literature:** Our search was limited to English-language sources to reflect the language competency of the reviewers. An electronic bibliographic search in EMBASE and MEDLINE was conducted. A discussion at the beginning of the study determined the inclusion and exclusion criteria and it was updated as the study progressed. Table 1 lists the medical subject headings (MeSH) terms that were used.

**Primary review:** Two investigators (KC, SH) independently reviewed all documents and completed the data abstraction. The differences were resolved by two third investigators with extensive international surgical experience (DP, LCL). Inclusion criteria were as follows: plastic surgery focus, pediatric population, global health perspective, within 2000-2013. Articles needed to meet all inclusion criteria. Items were excluded if less than 20% of pediatric patients were involved, did not have a global health focus, was not in English or was not able to be retrieved.

## Secondary review – Data charting, collating, summarizing and reporting

Prior to the secondary review, four investigators (ED, CM, LCL, DP) were provided with a sample of articles related to their field of clinical interest. Investigators identified the themes found in that sample. These themes were used in the secondary review by three investigators (KC,ED,CM). Themes were updated as the study progressed. Descriptive demographic characteristics were also collected at this time.

**Data analysis:** Analyses were performed only on the data from included studies. Demographic data, themes and research needs were summarized using basic descriptive statistics (simple counts and percentages).

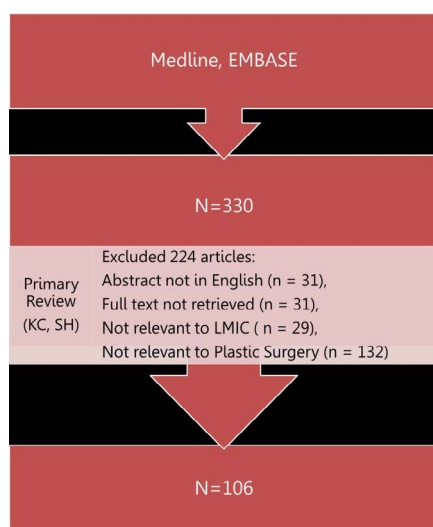
## Results

Figure 1 provides a flow diagram of the screening process. Only 106 studies satisfied the inclusion criteria after the initial literature review of 330 citations. Study demographic characteristics are summarized in Table 2. Twenty-six percent of the articles were authored by international agencies (n=28), with the majority of articles published from local institutions (n=78). Themes and research needs identified for Burns, CLP and International Agencies are summarized in Figure 2.

**Burns:** Publication distribution by continent is as follows: Asia (60%), Africa (33%), and Central/South America (3%). Local institutions published 94% of the publications. Table 3 summarizes the distribution of publications by country. Sixty percent of burn publications focused on burn epidemiology. Burns in males were more common than in females. Of the total number of articles that reported incidence (n=32), 28% reported higher burn incidence in females. Common risk factors included lower socioeconomic status, lower education, and crowded conditions [21-24]. Scald injury was reported to be the highest incidence for burns [6,24-26]. Fifty percent of publications indicated that children are at higher risk for burns than

Table 1. All search terms used for Medline and Embase.

Subject of Review	Scope of the term
What	Plastic Surgery, Plastic Surgical Nursing, Cleft*( cleft lip/ or cleft lip face palate/ or cleft lip nose/ or cleft lip palate/ or cleft palate/) Craniofacial*( craniofacial development/ or craniofacial malformation/ or craniofacial surgery/) Burn* Hand*(hand injury/ or hand joint/ or hand malformation/), Graft* ( full thickness skin graft/ or composite skin graft/ or free tissue graft/ or muscle graft/ or tissue graft/ or graft dysfunction/ or acute graft versus host disease/ or acute graft rejection/ or split thickness skin graft/ or nerve graft/ or free skin graft/ or graft failure/ or skin graft/)
Who	Pediatric* (Pediatric/ Pediatric Surgery/ Pediatric Ward) Child <unspecified age> Infant <to one year> Preschool Child <1 to 6 years> School Child <7 to 12 years> Adolescent <13 to 17 years>
Where	LMIC (developing country/ lowest income group/ international cooperation/ public health/ rural health care/)
When	2000 – 2013
Subject of Review	Scope of the term
What	Plastic Surgery (Surgery, Plastic) Cleft* (Cleft Palate/ Cleft Lip) Craniofacial*(Craniofacial Dysostosis/ Craniofacial Abnormalities) Burn*(Burns, Chemical/ Burns/ Burns, Electric) Hand* ( Hand Deformities/ Hand) Graft* (exp surgical flaps/ or exp free tissue flaps/ or exp perforator flap)
Who	Pediatric* (Pediatrics/ OR Child (adolescent/child/ child, preschool/infant)
Where	Poverty* (Poverty/ Developing Countries) International Agencies (International agencies/ red cross/ united nations) Rural Health (Rural Health)
When	2000 – 2013



**Figure 1.** Flow Diagram of Search Strategy and Screening Process: Overview of inclusion criteria, exclusion criteria and final distribution of articles.

**Table 2.** Demographic summary of each clinical domain based on year, location, institution type and publication type.

Type of demographic	Number of Studies	Percentage of Studies
<b>Publication Year</b>		
2000 – 2007	63	48%
2008 – 2014	69	52%
<b>Clinical Domain</b>		
Burns	50	47%
Cleft Lip/Palate	32	30%
General (Greater than one domain)	10	9%
Craniofacial	6	6%
Flaps/Grafts	3	3%
Hand	1	<1%
No Clinical Focus	4	4%
<b>Study Location</b>		
Asia	62	45%
Africa	42	31%
Central/ South America	12	9%
Other	11	8%
Not Stated	10	7%
<b>Study Institution</b>		
International Agencies	25	24%
Local Institutions	81	76%
<b>Study Design</b>		
Retrospective Cohort	43	36%
Editorials/ Letters/ Opinion Pieces	18	15%
Epidemiological Studies	12	10%
Case Reports	11	9%
Qualitative Research	11	9%
Prospective Cohort	8	7%
Audits	7	6%
Case-Control	5	4%
Reviews	4	3%

adults, where the highest incidence in children occurred in those aged 0-5 years (n=17). Burn mortality ranged from 27-53.5% [27,28]. Ideal management included carefully planned protocol [8]; immediate care [29]; open exposure, letting the burn dry out under the skin [24]; seven-flap-plasty for contractures [30] and; a figure-of-8 sling for post burn axillary contractures [31]. Sixty-four percent of articles cited a need

for burn prevention research and fifty-four percent of articles reported a need for public education. Effective burn prevention methods published included targeting those at higher risk, comic books for pre-school children and local, culturally-sensitive workshops and public education programs [26,33-35].

**Clefts:** The majority of cleft articles are published in Asia (47%), Africa (41%), then Central and South America (9%). International agencies published 29% of the articles (n=12). Table 4 summarizes distribution by country. Sixty-six percent of articles focused on epidemiology and thirty-eight percent reported on the significance of multidisciplinary identified risk factors for cleft lip/palate (CLP, 38%). Cleft lip/palate incidence in Asia ranges from 1.1 to 4.04/1000 live births [12,13,36,37], Africa 0.18/1000 to 5/1000 [37,38], the highest reported in the Wudoaba community of Ghana [37], and Central America from 1 to 2/1000 [39,40]. Mean age of repair ranged from 3.7 months [37] to 9.9 years [41]. Age of presentation ranged from zero to sixty-two years of age [36,41-44]. Cleft incidence in males is more common than in females, with left unilateral cleft lip occurring more frequently [41-44].

Thirty-eight percent of articles discussed preventative and risk factors. Preventative factors include folic acid and other micronutrients [40,45]. Risk factors reported include genetics, low socioeconomic status, teratogenic agents, nicotine poisoning, and exposure to organic solvents, radiation and alcohol [40,44,46]. Genes identified include Msh homeobox 1, forkhead box protein E1, and interferon regulatory factor [39].

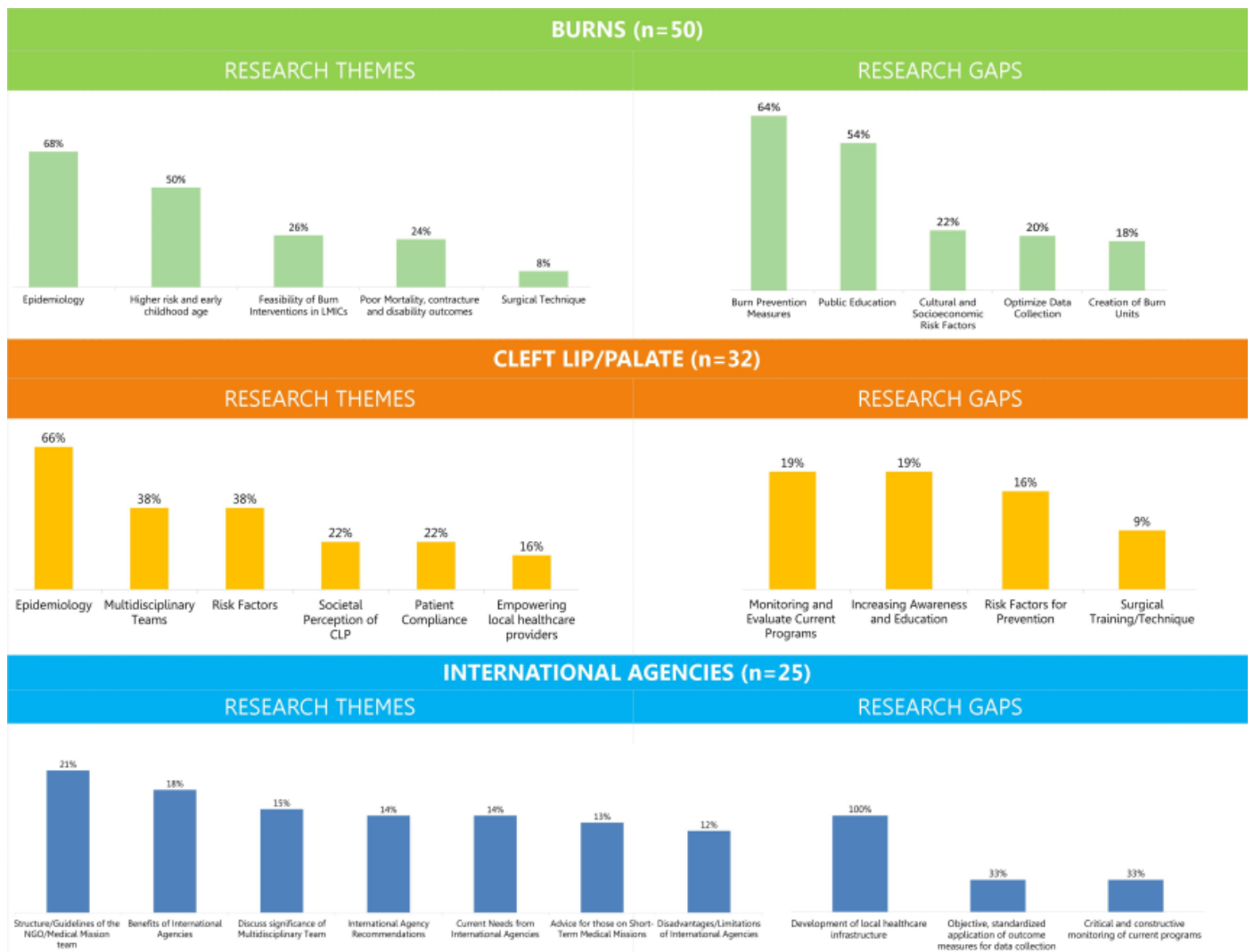
The dominant research gaps identified were: critical and constructive monitoring and evaluation of current cleft programs (19%), increasing public awareness and education (19%), targeting risk factors for cleft prevention (16%) and further discussion on appropriate surgical technique (9%). Interventional trials that target CLP risk factors are encouraged, specifically one that applies the use of folic acid [40,45]. There is indication that early repair of both lip and palate in one surgical session is not inferior in outcome to the traditional staged procedure [11]. Millard and Tennison have been cited as favored techniques for cleft lip, and Langenbeck-Wardill or Dorrance techniques for cleft palate [39,47].

**General plastic surgery**

General plastic surgery articles emphasized environmental hazards to children (60%) and identified public education as a research need (20%).

**International agencies**

Twenty-four percent of the articles were authored by international agencies (n=25). The majority of publications came from international aid in Asia (44%). Table 5 summarizes the distribution of publications by International Agency. Multidisciplinary teams were emphasized. International Agency benefits included financial support [39,48,49], good quality surgical care [18,39,48-50,53], training [14,18,50] and empowerment of local communities [14,18,39,50,52] (n=19). Recommendations included incorporating local leadership [14,15,17,36,51], teaching [14,17], sustainable follow up [17,39,52]. Standardized monitoring and evaluation [17,39,53,54] (n=15). Limitations cited from these publications included interfering with the local healthcare system, selection of sites based on volunteer preference rather than site need [14,15], lack of appropriate follow up and monitoring [36,55], and the infamous ‘body count [14-16,39,49] (n=10). Articles identified the following as a research need: public education [14,17,48,55], development of sustainable local healthcare



**Figure 2.** Major Research Themes and Gaps: Summary of major research themes and research gaps for Burns, Cleft Lip/Palate and International Agencies. Research gaps refer to the needs identified by the publications.

infrastructure [14-18,36,39,50-52] and standardized monitoring and evaluation (n=10) [17,39,53,54].

**Discussion**

The majority of research articles came from Asia and Africa. It appears that local institutions in India, South Africa and Bangladesh have been producing the most research. This also demonstrates that they have centers with appropriate infrastructure for research activity. These higher resource centers can consider building a foundation in prospective studies, randomized control trials or even experimental labs using local resources, based on identified research gaps. An example for a project could be instituting injury prevention measures in a small community and testing the incidence of traumatic injury thereafter. This will likely require international partnerships to successfully complete and may be an effective direction of resources. These centers also have the opportunity to collaborate with centers that have no research activity or fewer publications within their country. There is an obvious need for research articles from Central/South America, and LMICs from other continents not previously mentioned. Reasons for this are speculative, but epidemiological research to identify prevalence would be an appropriate start.

The majority of publications in global pediatric plastic and reconstructive surgery focus on burns and cleft surgery/pathology, provide the foundation for a systematic review and meta-analysis of literature in pediatric burns and cleft research in LMICs. In low-resource settings, research comes at an additional cost to already limited and stretched supplies and resources. However, an interesting difference in the two areas focuses on who is studying the condition. A vast majority of burn research is published by local practitioners (94%) rather than international agencies. While the majority of cleft lip/palate articles are also published by local practitioners, it is a much smaller majority at 62.5%. The relative abundance of local interest in burn pathology related articles suggest that local practitioners find it worth focusing their efforts to study and report this. This may be because the care for burn wounds is more complex, takes longer and has a significant impact on appearance and function. In comparison, local cleft related literature is bolstered by international or resourced interest. It may be due to better marketing from well-established agencies that have engendered sympathetic donors, providing a wealth of resources for both provision of care and education.

Another interesting finding is that both the burns category and the general plastic surgery category focus on injury prevention as a need.

**Table 3.** Number of Burn Publications Distributed by Country.

Location	Organization	Number of Publications
India	Chistian Medical Hospital, LokNayak Hospital,Forensic Medicine Hospital, Multi-centre; KanchiKamakoti CHILDS Trust Hospital, Government Medical College	7
South Africa	Edendale Hospital; Red Cross Children's; Chris Hani Baragwanath; UNISA Institute, Medical Research Council, Karolinska Institute	7
Bangladesh	Karolina Institute; Faridpur Institute; Multi-centre	6
Pakistan	Aga Khan University Hospital; Aman Hospital;Multi-centre	4
Iran	Department of Public Health; Tehran University of Medical Sciences	3
Sri Lanka	Multi-Centre, National Hospital of Sri Lanka	3
Egypt	Multicentre; Ain Shams University	2
Kenya	Kenyatta National Hospital; Kijabe Hospital	2
Nigeria	Lagos State University; Ahamdu Bello University Teaching Hospital	2
World Database	Karolinska Institute; Prince of Wales Hospital	2
Afghanistan	Hospital (Red Cross)	1
China	People's Liberation Army Hospital	1
Iraq	Red Cross	1
Malawi	Kamzumu Central Hospital	1
Mozambique	Multi-centre	1
New Zealand	Middlemore Hospital	1
Oman	Khoula Hospital Burns Unit	1
Peru	National Institute of Child Health Hospital	1
Sao Tome and Principe	STP Central Hospital	1
Sierra Leone	Multi-Centre	1
Singapore	Raja IsteriPengiranAnakSaleha Hospital	1
Sudan	Elfasher& Khartoum Bahr Teaching Hospital	1
Turkey	Baskent University Adana Hospital	1
Vietnam	BG UnfallklinikLudwingshafen	1
Yemen	King Faisal University	1

**Table 4.** Number of CLP Publications Distributed by Country.

Location	Organization	Number of Publications
India	Multi-centre; Operation Smile; Smile Train Project; Sri Ramachandra University	5
N/A	Multi-centre; Operation Smile	4
Nigeria	Lagos State University and Hospital; Author Specific	3
Bangladesh	Multi-centre	2
Cambodia	Children's Surgical Center	2
China	Operation Smile; University of Hong Kong	2
Ghana	KomfoAnokye Teaching Hospital	2
Africa	Multi-centre	1
America, Argentina, Guatemala	Rotaplast International	1
Brazil	Interplast	1
Columbia	Healing the Children	1
Democratic Republic of Congo	Doctors on Call for Service Learning Centre (DOCS LC) of Goma in the DRC	1
Iraq	Operation Smile	1
Mexico	Author Specific	1
Nepal	Author Specific	1
Vietnam	Japanese Cleft Palate Foundation	1
Wales (low SES)	Author Specific	1
Zambia	Zambia University Teaching Hospital	1

While the exact environments of small children are quite varied across the globe, it is clear that children are susceptible to injury. This type of research therefore, would need to be site-specific, with investigations into the direct causes of injury and the development and study of locally applicable interventions.

The epidemiology and prevalence of CLP is more established with clear variation within continents. Identification of higher incidence areas, such as that in Ghana's Wudoaba community discovered lead to more effective direction of future research and intervention to improve outcomes. A controlled trial or prospective review exploring interventions that target CLP risk factors in LMICs is highly

recommended [40,44,46]. Another research gap emphasized for cleft lip and palate is the monitoring of current programs. A prominent concern is that the ethics and safety of these missions should be evaluated. Quality improvement a vital aspect of most hospitals in high-income countries, and it is refreshing that there is a concern that charity programs need to be viewed critically as well. This theme is echoed by a more specific review of the articles published by international agencies [56,57].

The substantial research deficiencies in craniofacial defects, flaps, grafts and pediatric hand surgeries compared to pediatric burns and cleft lip/palate may direct future allocation of resources. The paucity

**Table 5.** Number of Publications Distributed by International Agency.

Organization	Location(s)	Number of Publications
Operation Smile	Nicaragua, Iraq	5
ReSurge/Interplast	China, Sri Lanka	4
Smile Train	India	2
Volunteers in Plastic Surgery	N/A	2
A Thousand Smiles	Mexico, City, Mexico	1
Facing the World	UK	1
Healing the Children	Columbia	1
International Noma Project	Sokoto, Nigeria	1
Japanese Cleft Palate Foundation	South Vietnam	1
MACLA: Medical Aid for Children of Latin America	Bolivia	1
Northern Cleft Foundation	India	1
Smile China	China	1
Sterling Bunnell Traveling Fellowship	Honduras, Cambodia	1
University of Insubria, Varese, Italy	Bangladesh	1
World Craniofacial Foundation	India	1
Yale School of Medicine	Manaus, Brazil	1

of pediatric hand surgery research in global health is significant and is likely under-reported given the high incidence of trauma in developing countries. This warrants further investigation because of the lifelong handicap for children with severe hand injuries. It would be worthwhile to investigate the epidemiology of pediatric hand surgery cases in LMICs to establish need.

Development of sustainable local healthcare infrastructure is emphasized as an area for future research for international agencies [14]. Quality evaluation and assessment are essential in order to maximize the care provided to vulnerable children in resource-poor situations [16,17,19,58]. Invested international agencies have recently begun to report on the quality of care provided. To maximize care quality, Volunteers in Plastic Surgery (VIPS) has developed pediatric plastic surgical guidelines to be used as a framework for short-term surgical missions [56,57].

### Limitations

First, the scope of the literature available likely does not reflect the extent of global need because of deficiencies in infrastructure, training, follow-up and financial support needed for local surgeons to report the activities at their hospital. Although this emphasizes the need for this review to streamline future research efforts, readers should be aware that other research gaps likely exist. Second, although this review can be cited as an international review, the scope was unfortunately limited to articles published in English. Future international research collaborations inclusive of other languages is recommended. Third, the depth of this review was also limited due to the very nature of the scoping review methodology. This compromise was needed in order to focus on mapping prevalent research themes and gaps. A focused systematic review in a more homogenous field such as burns or cleft lip/palate would be worthwhile. Lastly, a significant number of articles were not able to be retrieved because of limited access to specific journals which included those not published in English.

### Conclusions

The majority of publications in global pediatric plastic and reconstructive surgery focus on burns and cleft surgery. Epidemiological research has been well-reported in these domains and it would be worthwhile to conduct experimental and clinical trials in these fields. Clinical domains that are likely under-reported include pediatric hand

trauma, the use of flaps and grafts and other craniofacial anomalies. Epidemiological research from Central/South America would be welcome. Emphasis on developing local infrastructure and evaluating current international volunteer missions is appreciated. The significant gaps remaining in global pediatric plastic/reconstructive surgical research constitute opportunities for future international projects. The authors would be happy to provide all the references included in this scoping review to promote international collaboration in streamlining research efforts.

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### Conflicts of interests

None.

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